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Case Report

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Solitary Bone Metastasis as Initial Presentation in a Young Lady Diagnosed with Ovarian Carcinoma without Peritoneal Disease and Distant Metastasis

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Abstract

We are reporting a case of a 29 year old female diagnosed with ovarian carcinoma who initially presented with a single bony metastasis without peritoneal disease and no other distant metastasis. A single lytic lesion in the pelvic bone without any disease in the body except an ovarian tumor in a young female is an extremely rare case. The order of ovarian carcinoma spreading to distant organs are as follows i.e liver, distant lymph nodes, lung, bone and brain. Bone is considered the 4th most common site of metastasis. Worldwide and in India, ovarian cancer ranks at 8th position and the 5 year prevalence is in increasing trends. As the stage advances, the survival decreases and the prognosis becomes poor. Near about 60 % of ovarian carcinoma patients are diagnosed with advanced disease with distant metastasis at initial presentation. There is no streamline guidelines for these patients as lack of prospective data with availability of only retrospective case series. Aggressive treatment measures are necessary for treating ovarian carcinoma patients with bone metastasis. Quality of life of patients will be affected if skeletal related events occurred. Hence, comprehensive treatment planning will be helpful in improving the survival time and quality of life of patients of ovarian carcinoma with bone metastasis.

Keywords: Ovarian cancer; Bone metastasis; Distant metastasis; Young lady; Peritoneal disease; Prognostic factors.

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Introduction

Ovarian cancer is the 8th most common cancer, and the 8th most common cause of death from cancer in women worldwide. The risk of ovarian cancer is highest in high-income countries (8.2 Age Standardized Ratio per 100,000 versus 4.7 in low-income countries) but is rising in lower-income countries as they develop economically [1]. Number of new ovarian cancer cases in all age groups and number of deaths due to ovarian cancer in all age groups in 2020, worldwide are 3,13,959 and 2,07,252 respectively [2]. In India, ovarian cancer ranks at 8th position with incidence 3.5%, mortality 3.8% and 5-year prevalence around 15.45 % in all age groups [3]. The 5 –year survival rate of all types of ovarian cancer for women is around 47-50% [4]. The mortality rate is high for ovarian cancer as most of the cases are diagnosed at an advanced stage. It is reported that around 60% of ovarian carcinoma patients are diagnosed with advanced disease with distant metastasis [5]. Ovarian carcinoma metastasize to distant organs orderly as follows i.e. liver, distant lymph nodes, lung, bone and brain. Thus, bone was reported to be the 4th common metastatic site [6]. The literature lacks the screening guidelines for bone metastasis in ovarian cancer. Absence of early symptoms in bone metastasis delays the diagnosis and further treatment procedure [7]. Skeletal related events will occur in case of advanced bone metastasis which includes pain, malignant hypercalcemia, pathological fracture, spinal cord compression and bone marrow aplasia. All these events are responsible for worsening the quality of life of patient [8,9]. Being bone metastasis is the fourth most common site in ovarian carcinoma, the overall prognosis is poor. There is a need of prospective randomized database of bone metastasis in ovarian carcinoma which will define the risk factors and prognostic factors.

Case report

A 29 year old young female with no co-morbidity with Eastern Cooperative Oncology Group Performance Status 1 (ECOG PS- I) presented with generalised, vague pain in abdomen for the last 4 months. She had consulted an orthopaedic surgeon for the right hip joint pain 4 months back. She had been advised X –Ray pelvis. She didn't go for X-ray and received analgesics. After 2 weeks, she had dull aching pain in the abdomen. Again, she neglected it and continued analgesics for the next 2 months. Pain was not subsided with analgesics, thus she consulted a gynaecologist and she had been advised ultrasound of abdomen and pelvis. Ultrasound was suggestive of right adnexal lesion of size 17 x 15 x 10 cm. She had been referred to our clinic with this report. Clinical examination was suggestive of large palpable abdomino-pelvic mass of size approximately 18 x 16 cm which was mobile with well defined upper border with non-palpable lower border with free fluid in the abdomen. She had undergone laparoscopic left ovarian cystectomy 5 years back with no available documentation of previous histopathology. No relevant family, past and medical history. She was married at the age of 20 years and she had two children, one son, one daughter, both were full term normal deliveries. Per vaginal and per rectal examination was unremarkable. In view of right hip joint pain and right adnexal lesion, we had advised her PET–CT (Positron Emission Tomography –Computed Tomography) and tumor markers i.e CA 125, CEA, CA 19.9. PET was suggestive of right adnexal, solid–cystic lesion of size 17.2 x 16.2 x 9.9 cm with nodularity over omental surface with multiple enlarged lymph nodes at bilateral pelvic region and retroperitoneal region with single osteolytic lesion with SUV max 8.6 in the right iliac bone (Figure 1,2). CA 125 was 71 U/ml and rest of the tumor markers were within normal range.

Case was discussed in our Institutional Multidisciplinary Tumor Board. Board's decision was to start with neo-adjuvant chemotherapy with curative intent in view of oligometastatic disease. Hence, planned a procedure of diagnostic and staging laparoscopy with biopsy from the omental deposit or peritoneal deposit (As per PET findings). She underwent diagnostic laparoscopy. There was no disease in the peritoneal cavity. No disease or deposit over omentum, liver, spleen, bilateral subdiaphragmatic peritoneum, parietal peritoneum, bilateral paracolic gutter and bowel serosa. Minimal free fluid was present in the hepato-renal pouch. Ascitic fluid was sent for cytology. The only positive finding was right ovarian disease with normal left ovary. No suspicious tissue was available for biopsy. Ascitic fluid cytology turned out negative for malignant cells. Case was re-discussed in the board and considering her age, plan was changed to exploratory laparotomy with excision of right adnexal tumor with frozen section control followed by primary cytoreductive surgery as there was no peritoneal disease. She had been posted for surgery after getting fitness from anesthesist and physician. She underwent exploratory laparotomy. Right salphingo-oophorectomy specimen sent for frozen section and it was reported as high grade serous ovarian cystadenocarcinoma with adjacent areas having features of borderline ovarian tumor (Figure 3). Hence, we proceeded with primary cytoreductive surgery i.e. total abdominal hysterectomy with bilateral salphingo-oophorectomy (Figure 4) with bilateral pelvic lymph node dissection with para-aortic lymph node dissection with complete omentectomy. Oral feeding was started 12 hours post surgery. Postoperative course was uneventful. She was discharged on 7th postoperative day.

Final histopathology report demonstrated a high grade papillary serous cystadenocarcinoma of right ovary. Left ovary, uterus and fallopian tubes were normal. Bilateral pelvic lymph nodes (right - 0/11, left -0/12) and para-aortic lymph nodes (0/11) were free from metastasis with no deposit over omentum. AJCC 8th edition pathological staging was Stage IVB (p T1a p N0 p M1a). She had been advised adjuvant chemotherapy with 6 cycles of carboplatin and paclitaxel followed by radiation to right iliac bone. After 3 weeks of surgery she had been challenged with chemotherapy. She had tolerated chemotherapy without any major adverse effects. After 2 weeks of chemotherapy, she had received radiation therapy. She is in close follow up with us. After 6 months of treatment, PET CT was advised which showed no residual disease or new finding. After completion of one year post treatment, she is still disease free and she has been advised regular follow up as per our institutional follow up protocol.

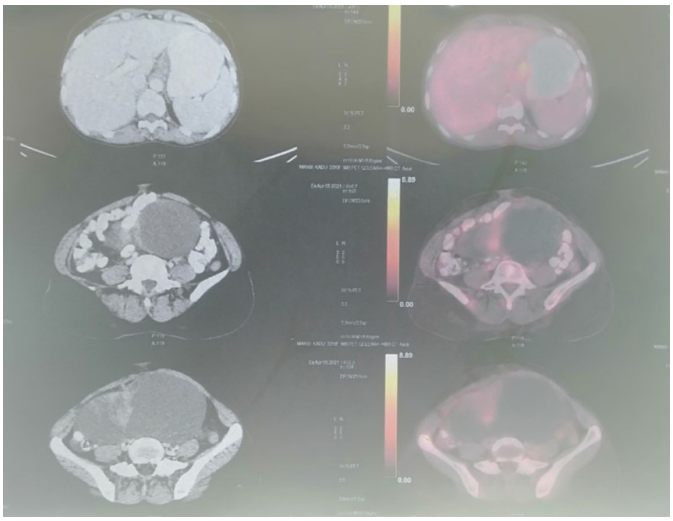

Figure 1: PET images of right adnexal lesion

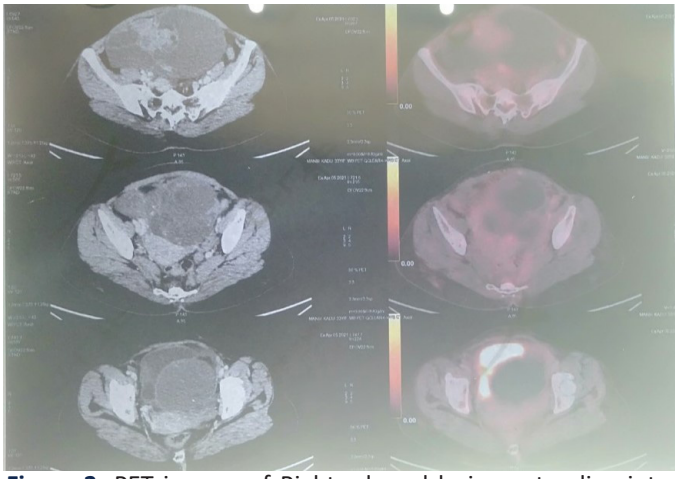

Figure 2: PET images of Right adnexal lesion extending into pelvis.

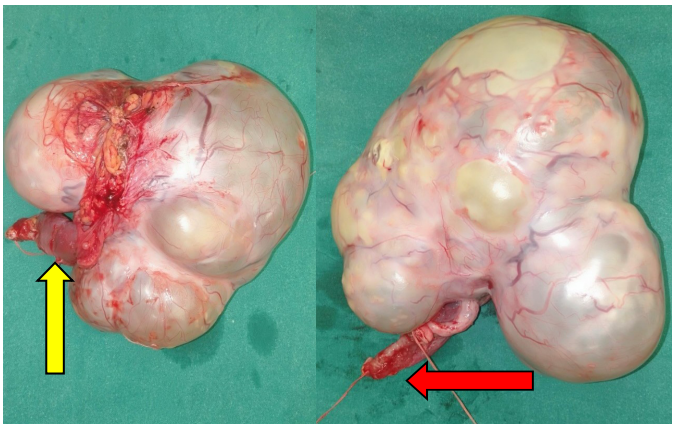

Figure 3: Right ovarian tumor (Ovarian vessels – marked by yellow arrow, fallopian tube – marked by red arrow)

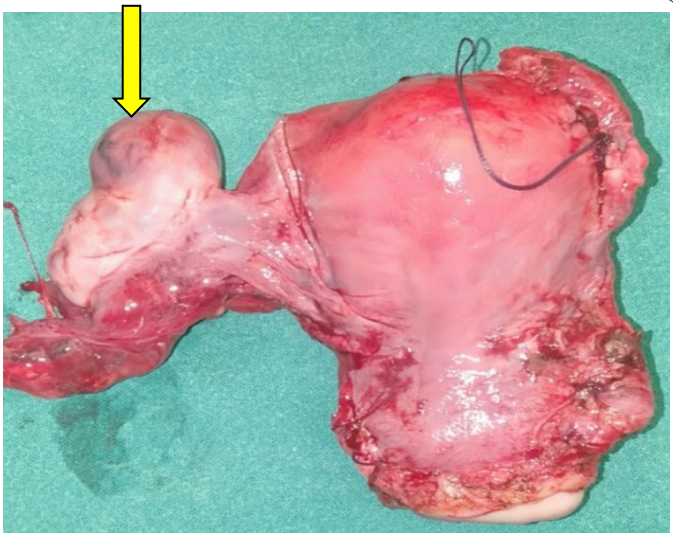

Figure 4: Hysterectomy specimen (Left ovary – marked by yellow arrow).

Discussion

Ovarian carcinoma usually spread to bone directly, hematogenously, via lymphatic spread or, transperitoneally [10]. The vertebral venous system plays the vital role in spreading ovarian cancer to bones. Several case studies have reported that the most common targeted bones which are susceptible for metastasis from primary ovarian cancer are pelvic bone and vertebral column [11]. Median survival of patient with bone involvement is very less. Some of the case series had reported the median survival of patients with bony metastasis around 7.5 months [12]. Multiple studies were published where different bone involvement as a metastatic site had been described such as Baize et al [13], described involvement of left iliac ramus as metastases from primary ovarian cancer, whereas Tiwari et al [14]. demonstrated a bone metastases in lumbar spine, while Abdul-Karim et al [15], reported bone metastases in seven patients with the involved bones are thoracic vertebra, clavicle, and axial skeleton [15]. In the index case, there was only a single lytic lesion in the right iliac bone. To avoid skeletal related events which occurs because of bony metastasis, bone targeted agents are necessary which helps in reducing bone resorption and decreasing the skeletal complications [16,17]. Denosumab is a human monoclonal antibody which is useful for reducing bone loss in malignant diseases [18].

Identification of risk factors is very important in planning, guiding and screening of treatment. Several case series were published in which association of risk factors with bone metastasis were demonstrated. One study had described, poor differentiated grade, lymph node involvement and advanced age were strongly associated with development of distant (Bone) metastasis [6]. Zhang et al [19], extended the research and described presence of older age, T2/T1 stage, N1/N0 stage, non-serous histology and the presence of lung and liver metastases are associated with de novo bone metastasis. There is still controversy between histological type of ovarian cancer and bone metastasis. Abdul-Karim et al [15], reported that high grade ovarian carcinomas are associated with bone metastasis instead of low grade carcinoma. Julian et al [20]. reported three cases of ovarian carcinoma with bone metastasis, out of which two were papillary serous carcinoma and one was with mucinous carcinoma. Zhang et al [19], demonstrated a strong association of non-serous ovarian carcinoma with

development of de novo bone metastasis and the median overall survival of ovarian carcinoma patients with bone metastasis was decreased to 5 months which is very less as compared to other reported case series where the median survival was reported to be approximately 8 months [21,22]. The study further described that the median survival time can be prolonged from 3 months to 18 months with primary tumour surgery. Hence, they concluded that aggressive surgery should be encouraged for ovarian carcinoma patients with bone metastasis and non-serous histology had negative association with the overall survival. In the index case, the histology was high grade serous carcinoma limited to one ovary with no peritoneal disease with no other distant metastasis except a single metastatic lesion in the right iliac bone and presentation in a young age. The index case contradicts to all available retrospective case series, hence we should consider the case as an extremely rare case with rarest presentation.

A multidisciplinary team approach is necessary for planning the treatment of ovarian carcinoma with bone metastasis. Multimodality treatment is require for treating the case. A comprehensive treatment is better than individualised treatment. The overall prognosis is poor with bone metastasis. The available treatment options are surgery, chemotherapy, radiation therapy, radionuclide therapy and bone strengthening agents. All measures should be utilized to improve the quality of life of patient. Till the availability of prospective randomized data, treatment guidelines cannot be implemented for all patients as it may differ from patient to patient. Thus, institutional guidelines and multidisciplinary board are the available best treatment options for ovarian carcinoma patients with bone metastasis.

Conclusion

Ovarian carcinoma with solitary bone metastasis with absence of peritoneal disease and without other distant metastasis is an extremely rare phenomenon and the overall prognosis is very poor in spite of all aggressive treatment measures. Comprehensive treatment planning will be helpful in improving the survival time and the quality of life of patients.

Disclosures

Human subject: Informed consent was obtained from the patient for being included in the study.

Conflicts of interest: The authors declare that they have no conflict of interests.

Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work.

Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

Availability of data and materials: Not applicable.

Consent for publication: An informed consent to publish this case was obtained from the patient.

Ethics approval and consent to participate: Not applicable.

Clinical trial transparency: Not applicable

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